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Partial Transvenous Coil Embolization with Significant Clinical Improvement in Patient with Indirect Carotid Cavernous Fistula

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ABSTRACT

Introduction: An indirect carotid-cavernous fistula (CCF) is an abnormal connection between the internal or external carotid artery and the cavernous sinus. The optic, trochlear, abducens, and trigeminal nerves are located in the cavernous sinus. Head Computed Tomography (CT) scan, Magnetic Resonance Imaging (MRI), and Cerebral Angiography are some imaging modalities used to establish and diagnose carotid-cavernous fistula. Endovascular intervention can be performed with trans-arterial or trans-venous access. In some cases, complete embolization by endovascular treatment is not possible because of difficult angioarchitecture. **Case:** A 61-year-old female reported having pain in her left eye (numerical rating scale was 6). The left eye was bulging, reddish, ptosis, and unable to move. The patient felt double vision when opening both eyes, complained of intermittent stabbing headaches on the left side, and heard a bruit from the left side of the head. The cerebral angiography showed bilateral indirect CCF Barrow type D. The procedure involved partial transvenous embolization with coiling. The angiographic evaluation showed partial occlusion, and residual flow from the fistula was still visible on angiography. There was a significant clinical improvement several days after the procedure and a five-month follow-up. **Conclusion:** This case report showed that partial targeted transvenous embolization is quite effective in indirect CCF when complete embolization is impossible.

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INTRODUCTION

A carotid-cavernous fistula (CCF) is an abnormal vascular connection between the internal or external carotid artery and the cavernous sinus.[1] The location of cavernous sinus is behind the eyes and receives blood from the brain, eyes, and pituitary gland. A carotid-cavernous fistula is classified as direct or indirect. CCF can be caused by trauma[2] or spontaneous.[3]

The cavernous sinus has essential structures, including the oculomotor, trochlear, abducens, and first division trigeminal nerve.[4] Fistula in the cavernous sinus can cause abnormalities in the eye, including eye injection, chemosis, proptosis, ophthalmoplegia, orbital pain, and bruits.[5] These abnormalities can cause clinical symptoms that disturb the patient's daily activities. Thus, CCF patients require immediate and optimum treatment.

Managing carotid-cavernous fistula is endovascular intervention, surgery, radiation, and manual vascular compression.[6] The treatment of choice is endovascular intervention. This case report discusses the indirect Barrow type D CCF, which improves clinically after a partial transvenous coil embolization procedure.

CASE

A 61-year-old female, in an outpatient clinic, with the chief complaint of left eye pain (numerical pain scale 6) for four months. The left eye was difficult to move, red, and swollen, followed by a closed eyelid. The visual acuity was within normal limits, but there was a double vision when looking with both eyes. The patient complained of intermittent left-sided headaches with a stabbing quality and heard a hissing sound inside her head, especially on the left side. She was disturbed and had trouble sleeping. It is known that the patient had a history of hypertension since five years ago.

Based on a general physical examination, the patient had high blood pressure. In the neurological examination, Glasgow Coma Scale E4V5M6, there were no signs of meningeal signs. An ophthalmology examination revealed visual acuity was >2/60, a positive light reflex in both eyes, and an isochoric pupil of 3mm/3mm. The fundoscopy test was within normal limits. There was binocular diplopia, left eye ptosis, left eye ophthalmoplegia, left eye pain, and bruit in the left supra orbit.

Head Computed Tomography (CT-Scan) with contrast, head MRI-MRA, and head angiography were performed. A contrast-enhanced head CT-scan revealed chronic lacunar infarcts in the left thalamus, oculi bulb, extraocular nodules, and optic nerves that were symmetrical right and left. There was no abnormal enhancement in the retrobulbar mass. MRI and MRA showed chronic lacunar infarction in the left thalamus, small vessel ischemia in the left frontal subcortex, and right-left lateral periventricular dilated superior ophthalmic vein (SOV) dilatation and early filling of the left cavernous sinus to the left inferior petrous sinus (IPS), suggestive of a carotid-cavernous fistula in the left orbit. Head angiography was performed, and it showed a bilateral indirect carotid cavernous fistula (CCF) of Barrow type D (Figure 2).

Transvenous coiling was performed on this patient. The catheter was inserted into the right and left external carotid artery (ECA) and the left sigmoid sinus. This intravenous embolization approach was targeted at the left cavernous sinus via the internal jugular vein to the inferior petrosal sinus (Figure 3). Followed by inserting two coils in the cavernous sinus to occlude the fistula. We performed an angiographic evaluation after the coils were inserted. The blood flow through the fistula was reduced but still found blood flow through the left meningeal accessory artery to the cavernous sinus (Figure 4).

One day after the coiling procedure, the left eye was still red, but the pain decreased (NRS 2), and the patient no longer heard the bruit sound from the left side of the head. One week after the coiling procedure, the headache was relieved, and pain around the eyes had reduced. There was swelling and no ptosis. The redness (chemosis) in the left eye disappeared. The left eye can move in all directions. There was no bruit sound. After five months of the coiling procedure, the patient said there were no complaints. The left eye was still slightly reddish, but it did not interfere with the patient's daily activities (Figure 1).

DISCUSSION

A carotid-cavernous fistula (CCF) is an abnormal connection between the internal or external carotid artery and the cavernous sinus.[3] Based on the flow, CCF is classified into a high and a low flow.[7] Meanwhile, referring to the imaging, it can be classified based on the Barrow classification. Type A is a direct high-flow connection between the internal carotid artery and the cavernous sinus, and Types B, C, and D are indirect low-flow connections.[7]

Several essential structures in the cavernous sinus include the oculomotor, trochlear, abducens, and trigeminal nerve.[8] Thus, the patient had abnormalities like ptosis, conjunctival chemosis, ophthalmoplegia, orbital pain, and bruits. These

clinical symptoms arise from disturbances in the cranial nerves that pass through the cavernous sinus.[9] The gold standard to diagnose CCF is head angiography. However, initial imaging often uses rely on non-invasive modalities such as a head CT scan or MRI/MRA.[10] From head angiography, found connections from the accessory meningeal artery, deep temporal artery, and ascending pharyngeal artery to the cavernous sinus on the right side. There were also connections from the accessory meningeal artery, deep temporal and ascending pharyngeal artery to the cavernous sinus on the left side. Thus, head angiography showed a bilateral indirect CCF Barrow Type D.

Management of the carotid-cavernous fistula itself can include endovascular intervention, surgery, radiation, and manual vascular compression.[6,11] The primary treatment for CCF is endovascular intervention.[12] The endovascular intervention is minimally invasive, can be performed without general anesthesia and has a faster recovery time after the procedure.[12]

The artery and cavernous sinus connection, direct or indirect fistule, are the basis for the endovascular intervention for CCF.[13] Endovascular intervention that can be performed for direct CCF is occlusion with a detachable balloon inserted via the arterial route to maintain the patency of the internal carotid artery.[14] A latex or silicone balloon is inserted through the microcatheter into the cavernous sinus via the femoral artery approach. In high-flow fistulas, the balloon was brought to the fistula location where the balloon will be inflated and released, resulting in fistula occlusion.[15] The success rate of trans arterial embolization for patients with direct CCF is 93.93% with complete obliteration.[13]

Endovascular intervention that can be performed for indirect CCF is transvenous coil embolization. The advantage of this procedure is that it can close the fistula from the outside artery if we cannot do trans-arterial access.[14] Venous access through the femoral vein inserted the catheter and then navigated into the cavernous sinus.[15] Previous case reports showed that the transvenous coil embolization procedure has different clinical symptom improvement. Successful rate of this procedure can be described as proptosis (89%), chemosis (92%), cephalic bruit (87.5%), tinnitus (100%), and diplopia (66.6%), N. III paresis (50%), N. VI paresis (50%), and visual loss (100%).[16]

In this case, due to the indirect fistula barrow type D, there were multiple connections between both ICA and ECA branches with cavernous sinuses,[12] we decided to perform transvenous coiling. We chose this procedure because the precise location and size of the fistula were challenging to identify. It was hard to perform a transarterial endovascular procedure.

Two catheters, one into the left sigmoid sinus and the other into the right and left ECAs, were inserted during the procedure. This approach to intravenous embolization was targeted at the left cavernous sinus via the internal jugular vein to the inferior petrosal sinus. Then, a coil was inserted to achieve venous occlusion. Based on head angiography showed decreased flow to the fistula, but there was still a small flow from the fistula through the left meningeal accessory artery. This result indicates that the occlusion was partial.

After the procedure, the symptoms of CCF usually disappear within a few hours to days. There was no visual impairment. Recurrence of CCF is rare, but it still needs to be followed up with a post-treatment angiography to examine the condition of the fistula.[17]

Another case report on bilateral indirect CCF was successfully treated with complete closure endovascular coil embolization.[18] This procedure showed spontaneous resolution of visual symptoms and immediate vision recovery.[18] In our case, we cannot close the whole fistula. After the postoperative angiographic evaluation, there was reduced flowing to the fistula, but the flow from the fistula was still visible through the left accessory meningeal artery. We planned to perform a second step embolization. However, there was a clinical improvement, such as reduced headache and eye pain. There was no ptosis, and the eyes could move in all directions. Ecchymosis is still occurred but reduced. She had normal visual acuity, and the bruit inside her head no longer listened. The partial transvenous coil embolization was beneficial and showed significant clinical improvement in this patient.

CONCLUSION

The bilateral indirect CCF Barrow type D case symptoms were ptosis, eye injection, conjunctival chemosis, ophthalmoplegia, orbital pain, and supra-orbital bruits. The gold standard for diagnosis of CCF was head angiography, and we chose an endovascular partial transvenous coil embolization approach for management; as a result, there was significant clinical improvement in the patient. This case report demonstrated that partial transvenous embolism is quite effective as an alternative therapy for indirect CCF type D.

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ATTACHMENT

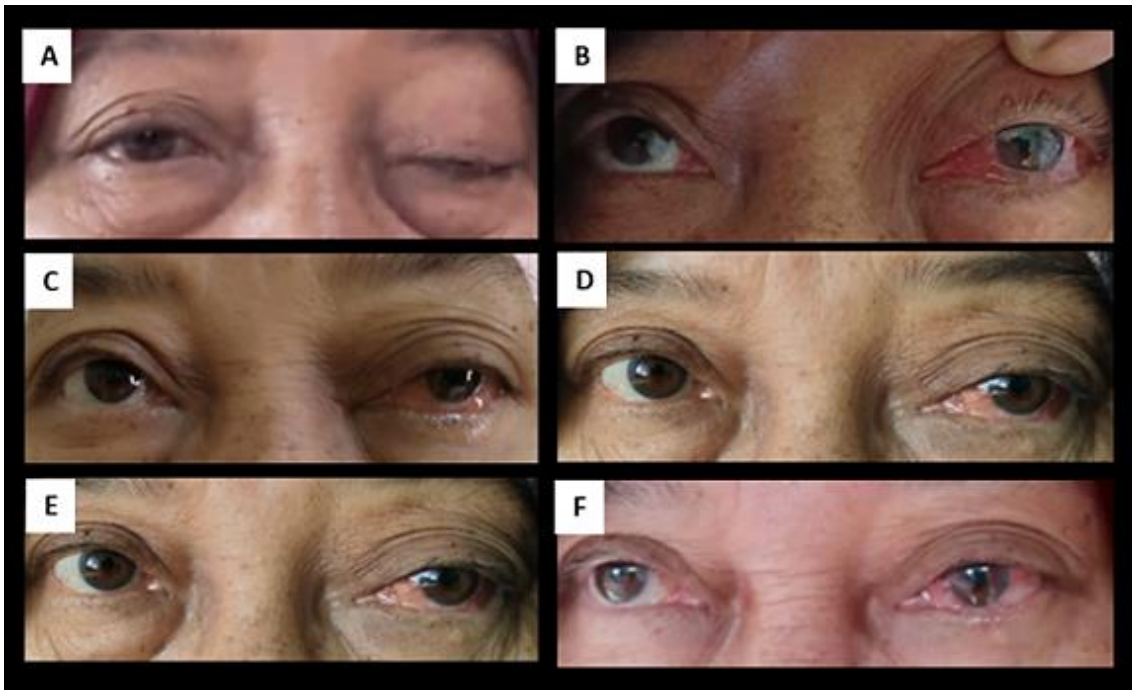

Figure 1. **Pre-coiling examination (A-B):** the patient had left ptosis, left eye injection and chemosis, left eye ophthalmoplegia, binocular diplopia, orbital pain, and supraorbital bruit in the left eye. **Post-coiling examination (C)** One day after partial transvenous coiling embolization, the complaints gradually decreased. **(D)** Five days after the procedure. **(E)** Fifteen days after the procedure, **(F) After five months after the procedure**, the conjunctiva of the eye is still reddish. No ptosis was found, eye movements were within normal limits, bruits and eye pain disappeared.

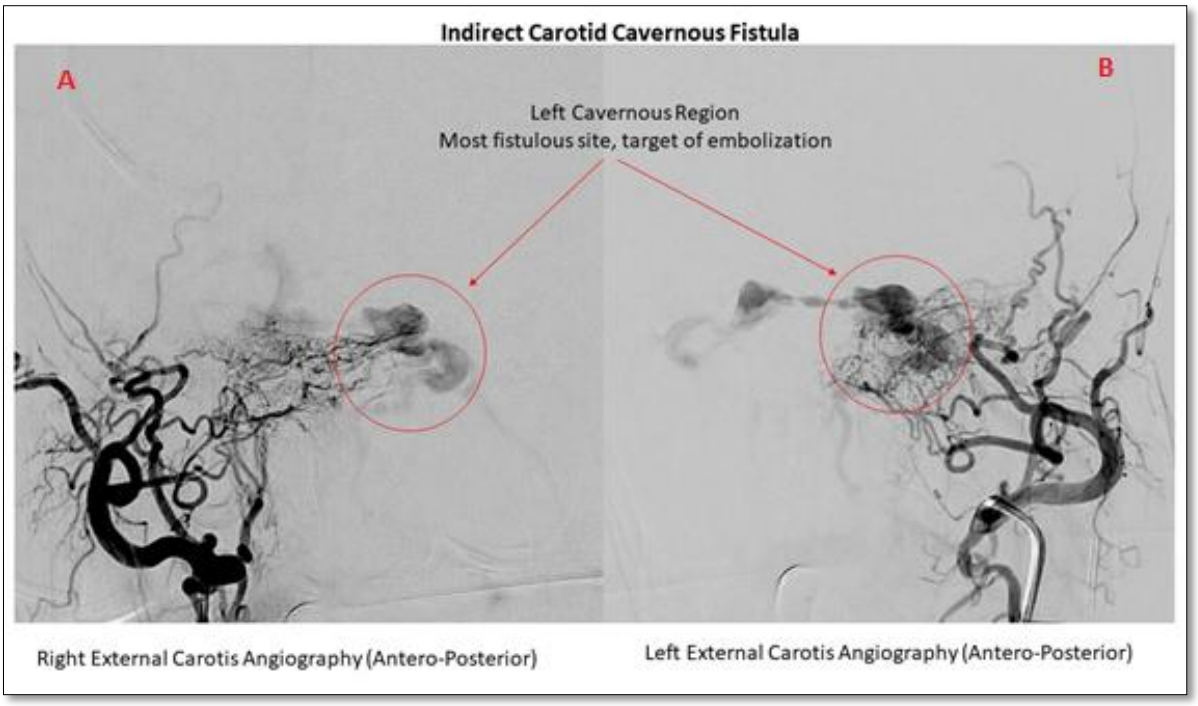

Figure 2**. A**. Contras was injected into the Right External Carotid Artery (RECA); connections from the accessory meningeal, deep temporal, and ascending pharyngeal arteries to the cavernous sinus. **B**. Contras was injected into the left external carotid artery (LECA), from the Accessory Meningeal, Deep Temporal, and Ascending Pharyngeal Arteries to the Cavernous Sinus. Cerebral DSA concluded that a Bilateral Indirect Carotid Cavernous Fistula (CCF) Barrow Type D was obtained.

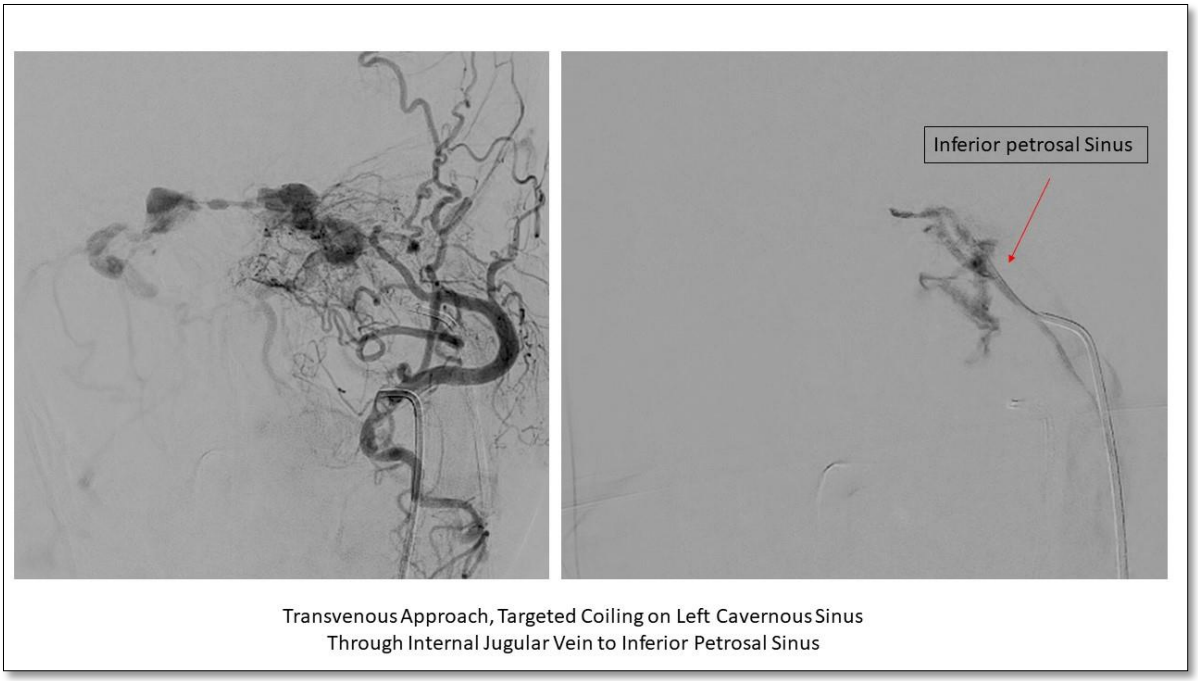

Figure 3. Transvenous embolization approach. Targeted at the left cavernous sinus via the internal jugular vein to the inferior petrosal sinus

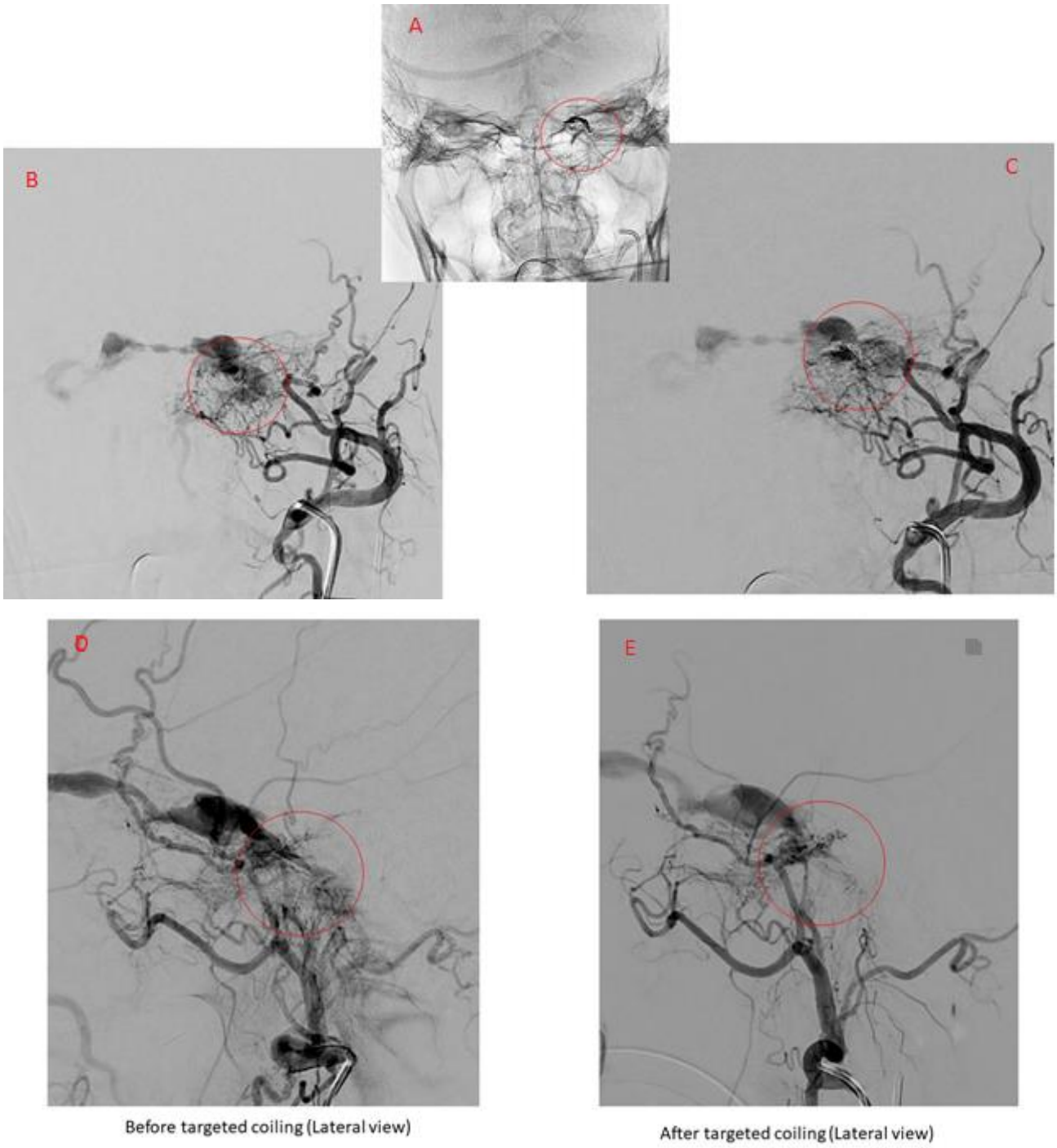

Figure 4. **A.** The coil seen after the procedure (anteroposterior view) **B.** Before coiling (anteroposterior view), **C.** After coiling (anteroposterior view). **D.** Before coiling (lateral view), **E.** After coiling (lateral view)